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Adaptation of the Quality of Life Adolescent Cleft Questionnaire for Spanish children and adolescents with cleft lip and/or palate

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Abstract

Background: The Quality of Life Adolescent Cleft Questionnaire is a measure of quality of life in cleft lip/palate (CL/P) including items on pre-treatment and post-treatment status. Items, however, were originally organized in a factor structure that prevents a formal pre-treatment to post-treatment comparison. Additionally, the questionnaire was tested in older patients.

Aim: We aimed to explore a factor structure that allows a comparison of pre- to post-treatment status in children and adolescents with CL/P.

Design: The sample comprised 60 children and adolescents with CL/P. The scale was divided into two groups of items (24 comparing pre-treatment and post-treatment status and 26 measuring current quality of life). Two different exploratory and confirmatory analyses were conducted (one for each group of items). Sources of criterion validity were investigated with measures of self-esteem and self-efficacy.

Results: The results supported a 6-factor structure for the pre-treatment and post-treatment items. In the second group of items, 9 items were removed due to inadequate functioning and a final 4-factor solution was obtained. The criterion validity of factors was good.

Conclusion: The proposed factor solution might be more useful to detect the perceived satisfaction in different areas and can be used in younger patients.

KEYWORDS

cleft palate, cleft lip, quality of life, factor analysis, validation study

1 | INTRODUCTION

Cleft lip/palate (CL/P) is one of the most frequent craniofacial malformations, affecting approximately 1 in 500 to 1000 births worldwide.[1,2] This malformation can give rise to a wide variety of aesthetic and functional problems, which have a profound impact on the physical and psychosocial functioning, as well as on the quality of life (QoL) of the individual.[2] Affected individuals have decreased facial growth, dental abnormalities, language disorders, hearing problems, and difficulties in psychological well-being and social relationships.[1] Thus, CL/P is associated with an increased risk of psychosocial problems, which include anxiety, social inhibition, depression, low self-esteem, externalizing problems, and attention-deficit/hyperactivity disorder.[2-4]

The incidence of CL/P in Spain is about 1.44 per 1000 live births,[5] with the most frequent pattern being the unilateral complete cleft lip and palate.[6] In Spain, cleft care is provided by regional hospitals with cleft team surgeons paid for by the Spanish National Health System. Dental and orthodontic care for patients with a cleft, however, is usually private, which makes accessing a sample with an adequate number of cases difficult, and therefore, there is little research on psychosocial variables in child patients with a cleft although analyzing quality of life among these individuals is a priority.[7]

Rehabilitation of CL/P requires multiple reconstruction surgeries that begin in childhood and continue into adulthood, aiming to correct the shape and function of the mouth and face,[2,8] including non-surgical orthopedics before lip seal surgery, long orthodontic treatments, speech therapy, and surgical insertion of pressure equalization tubes into the ears.[2,9] The interdisciplinary teams that carry out these extensive treatments are focused on achieving cleft closure, favorable facial growth, and adequate aesthetics and functionality, since the predominant objective is to improve psychosocial well-being and QoL in this population.[8]

Even though children and adolescents with CL/P are at high risk of experiencing behavioral disorders and social stigma, there is a shortage of specific and standardized instruments to assess psychosocial problems and health-related QoL (HRQoL) in this population.[2,10,11] Thus, it has been argued that the assessment of CL/P patients and their families might have been, to date, insufficient or inadequate.[3,12,13]

QoL depends on perceived health status and should be assessed by instruments that examine the impact of the disease and treatment modalities, while integrating both objective and subjective health indicators.[4] Particularly in CL/P, HRQoL should assess the direct impact of oral conditions on outcomes related to appearance, oral function, and social interactions, including an assessment of the patient's perception of treatment outcomes.[1,12] Examples of specific CL/P measures that assess at least some of these aspects of HRQoL are the CLEFT Questionnaire[9] and the Quality of Life in Adolescents with Cleft Assessment.[4] Importantly, although they both assess the status of patients after surgery and treatment, they do not measure the perceived changes in comparison with their status before surgery. This is important because in order to contribute to the existing literature showing that treatments focused on improving appearance and functionality appear to influence positively on the HRQoL of children and adolescents,[11,12] the evaluation of HRQoL should not only include current HRQoL but also perceived improvement compared with previous status, even if this is associated with some recall bias.[14]

Although there are previous studies on QoL in patients with cleft, generic instruments (eg, Child Oral Health Impact Profile-Short Form,[8] Child Perceptions Questionnaire[15]) have been used, which are less likely to be sensitive to all of the issues specific to the CL/P condition and the changes resulting from surgical treatment.[16] In contrast to the previous questionnaires, the Quality of Life Adolescent Cleft Questionnaire (QoLAdoCleft)[17] is a specific measure of HRQoL in CL/P populations that includes the assessment of perceived status both after and before treatments. The aim of this study was to adapt the QoLAdoCleft in a sample of Spanish children and adolescents with CL/P, and to make scale adaptations that would allow for a pre-post treatment comparison. It should be noted that, even though this is a promising scale for this population, its psychometric properties and feasibility of use across ages remain unclear. The original study describing the development and psychometric properties of the scale proposed a 3-factor solution (physical, psychological, and social health) and was validated in patients aged 16 to 24 years.[17] In relation to the former, however, it is important to consider that the psychometric soundness of the scale was solely based on the analysis of the internal consistency (Cronbach's alpha) of the three subscales and a general factor. Because no formal exploratory or confirmatory factor analyses were conducted, the extent to which the proposed 3-factor solution is the most appropriate to fit the data, as well as the utility of the general factor, remains unclear. Additionally, the 3-item structure did not differentiate items that referred to pre-treatment and post-treatment HRQoL status, so one factor contained both types of items together. This makes scores difficult to interpret and would make pre- to post-treatment status comparisons impossible. Finally, it would be of interest to carry out additional validations in younger ages due to an increased vulnerability at this stage.[18]

In addition to exploring the factor structure of the scale, we will also aim to study the sources of validity evidence by comparing scores with measures of self-esteem and self-efficacy. Self-esteem is related to an individual's set of feelings and thoughts about his or her self-worth,[19] and self-efficacy (defined as people's beliefs about their capabilities to produce the expected results[20]) plays a fundamental role in coping with life situations,[20] influencing the definition of goals, motivation, thought patterns, and emotional reactions,[21] and is an essential element that contributes to the well-being of adolescents.[20] High self-esteem combined with a sense of competence (self-efficacy) protects the individual from fear of isolation and social rejection.[22] Based on the above, a positive correlation of both variables with patient-perceived QoL is to be expected, which would support the construct validity of the instrument.

2 | MATERIALS AND METHODS

2.1 | Sample size and setting

Participants were 60 children and adolescents with a cleft. The type of cleft was specified in a questionnaire answered by the parents, and this information was cross-checked with the patients' medical records. This cross-sectional study was conducted among children and adolescents with CL/P who had undergone treatment at two specialized cleft centers in Madrid (Spain). No formal a priori sample size calculation was made. To determine our sample size, we considered the sample used by the original study where the QoLAdoCleft was developed (n = 40),[17] as well as data from previous research showing that sample sizes in studies into CL/P tend to be very small.[23] A minimum sample size of n = 60 was established, in accordance with the studies carried out in this population [24] including validation studies.[17] Data collection was performed between 2016 and 2018 and was obtained through self-reported questionnaires on paper that patients filled out during their orthodontic checkups. A researcher answered the participants' questions if they had any, without interfering with their answers. The inclusion criteria for the sample were to be aged between 8 and 18 years, having a cleft, being in orthodontic treatment, and having experienced previous surgery to improve their appearance and/or functionality. Patients with intellectual and developmental difficulties and syndrome-associated cleft were excluded. All patients were in orthodontic treatment and had undergone surgery to improve their appearance and/or functionality.

During the sampling period, 75 patients met the inclusion criteria, although only 60 (80%) completed the self-report questionnaires in full. A researcher personally handed out the questionnaires to the patients, in the presence of the parents, explaining the aim of the study. The main reasons for non-completion of the questionnaires were that parents, or the patients themselves, were not interested in participating. The exact reasons for the previous were not explored in detail.

The original instrument, provided by the author of the original study, was in English.[17] We conducted a double-translation/back-translation process and two consensus meetings. Two independent translators provided two target Spanish versions, which were translated back to English by two other independent translators. Translators and back-translators translated into their mother tongue. Discrepancies were solved by consensus, and we developed a back-translated English version, which was compared for equivalence with the original by a bilingual psychologist.

The study followed the guidelines of the research ethics committee of the Rey Juan Carlos University (110720166716), and informed consent was obtained from the patients' parents.

2.2 | Measures

In addition to the Quality of Life Adolescent Cleft Questionnaire,[17] which has been described above, we administered a measure of self-esteem (SEI) and a measure of self-efficacy (CPSE) to investigate sources of criterion validity of the QoLAdoCleft.

2.2.1 | Self-Efficacy

The Spanish validation of the Children's Perceived Self-Efficacy (CPSE) questionnaire, was used.[25] The CPSE contains 35 items rated on a 5-point Likert scale. This scale allows the assessment of three subdimensions: academic self-efficacy (alpha = 0.87), social self-efficacy (alpha = 0.81), and self-regulation (alpha = 0.78).

2.2.2 | Self-Esteem

The Spanish validation of the Coopersmith Self-Esteem Inventory (SEI) was used.[26] SEI contains 58 items and consists of 4 subscales: general self-esteem (alpha =0.79), school self-esteem (alpha =0.81), social self-esteem (alpha =0.83), self-esteem at home (alpha = 0.80).

2.3 | Data analysis

First, an analysis of item frequencies was conducted to explore the functioning of items. Analysis of kurtosis and skewness was carried out. Because many of the items, however, are focused on the status of patients after treatment, floor effects and therefore skewness and kurtosis problems were expected in items referring to post-treatment status.

Next, we divided the scale into two sets of items. The first group assessed pre- to post-treatment changes (24 items) and included pre-treatment items starting with 'Before orthopedic/orthodontic treatment…' and its corresponding version for the post-treatment 'At present…'. The second group of items assessed overall status without comparing current and pre-treatment HRQoL (26 items). Item distribution is indicated in Tables 1 and 2. An exploratory factor analysis was conducted with up to 6 factors for each of the two groups of items separately. More factors were only included if model fit was not sufficiently good with models with fewer factors, which are preferred for parsimony reasons. In the first group containing pre- to post-treatment items, the preferred structure was one that separated pre-treatment and their corresponding post-treatment items in different factors so that a pre- to post-comparison could be made. In the second group of more general items, the preferred factor structure

TABLE 1 Factor loadings of the proposed 6-factor model for pre- to post-treatment items with a confirmatory factor analysis

Item number and content	1	2	3	4	5	6
Before orthopedic/orthodontic treatment…						
1. Did you find difficulties in biting, eating and/or swallowing foods?	0.82					
2. Have you had difficulties in breathing?			0.60			
3. Have you suffered from ear pathologies?			0.60			
4. During meals, have you noticed any food reflux/regurgitation from the nose?	0.67					
9. Have you had difficulties in pronouncing a particular sound and/or word?	0.76					
10. Have your listeners had difficulties in comprehending your spoken language?	0.86					
13. Your voice was…	0.83					
15. Have you ever had any trigger points (painful points) limiting mouth and/or nose function?	0.78					
20. Have you ever felt different from others because of your cleft?					0.94	
21. Have you ever thought of yourself as less valid than others because of your cleft?					0.93	
24. Did you tend to isolate yourself because of your appearance?					0.83	
35. Has your physical and/or emotional status had any influence on your social activities?					0.87	
At present…						
5. Do you have difficulties in biting, eating and/or swallowing foods?		0.75				
6. Do you have difficulties in breathing?				0.51		
7. Do you suffer from ear pathologies?				0.65		
8. During meals, do you notice any food reflux/regurgitation from the nose?		0.55				
11. Do you have difficulties in pronouncing a particular sound and/or word?		0.72				
12. Do your listeners have difficulties in comprehending your spoken language?		0.91				
14. Your voice is…		0.93				
16. Do you have any trigger points (painful points) limiting mouth and/or nose function?		0.67				
22. Do you feel different from others because of your cleft?						0.85
23. Do you think of yourself as less valid than others because of your cleft?						0.92
25. Do you tend to isolate yourself because of your appearance?						0.88
36. Does your physical and/or emotional status have any influence on your social activities?						0.88

was based on item content and model fit. After looking at the item distribution in the exploratory models, a model was proposed and its fit was investigated by means of a confirmatory factor analysis. In the factor analyses, all items were labeled as categorical and the estimator used was the robust diagonally weighted least square mean and variance adjusted (WLSMV), which is specifically designed for categorical and ordinal data because it does not assume normality of distributions.[27] The usual fit indices were reported (ie, chi-square, RMSEA, CFI, and TLI). RMSEA values smaller than 0.05 and 0.08 are argued to reflect an excellent and a good fit, respectively. Similarly, CFI and TLI estimates over 0.95 and 0.90 are considered to reflect excellent and good fit, respectively.[28] As suggested in previous research,[29] only factor loadings equal or higher than 0.32 are presented as

these represent 10% of explained variance. The exploratory and confirmatory analyses were computed with Mplus version 6.12.[31]

After an analysis of the factor structure of the scale yielded an adequate structure, this was used to calculate the utility of the pre- to post-treatment scales and to investigate the sources of criterion validity of the questionnaire. Regarding the former, pre- to post-treatment scores were compared to confirm that the questionnaire, which was addressed after treatment, indeed captured perceived improvement in status after treatment by comparing pre- and post-treatment appraisals. Regarding sources of criterion validity, bivariate correlations were calculated between the QoLAdoCleft and measures of self-esteem and self-efficacy.

TABLE 2 Factor loadings of the proposed 4-factor model for non–treatment-related items with a confirmatory factor analysis

	1	2	3	4
26. Does answering to curious questions about your cleft embarrass you?	0.74			
27. Do you feel satisfied with your facial appearance?		0.87		
28. Do you feel satisfied with the aspect of your mouth?		0.96		
29. Do you think you need more surgical treatment to improve your facial appearance?		0.62		
30. Do you feel anxious about your cleft?	0.82			
31. Have you ever noticed any depressive symptoms related to your cleft?	0.85			
32. Are you worried about other people's thoughts regarding your pathology?	0.85			
33. Have you undergone psychological support therapy?	0.55			
34. Have you ever been bullied or been a victim of mockery because of your pathology?	0.51			
39. Are you satisfied with your family relationships?			0.62	
43. Do you find any difficulties in approaching your peers because of your cleft?	0.73			
44. Do you find any difficulties in approaching your colleagues and/or seniors because of your cleft?	0.83			
45. Do friends/colleagues/parents support your choices in managing your pathology?			0.76	
47. Are you surrounded by valid people with whom to share your worries and thoughts about your condition?			0.73	
48. Have you found any difficulties in accessing specialized centers for cleft care?				0.90
49. Have you found any difficulties in achieving adequate health care?				0.90
50. Have you found any difficulties in getting information about your condition from professionals?				0.98

3 | RESULTS

3.1 | Characteristics of the participants and previous descriptive analyses

The final sample was composed of 60 children and adolescents with a cleft (mean age =12.72, SD =2.73; range 8-18 years; 55% female). Ten participants had a cleft lip (16.7%), 14 had a cleft palate (23.3%), and 36 had a cleft lip and palate (60%). Ninety percent of participants had undergone at least two surgeries.

Before the exploratory analysis, we analyzed the frequency tables for all items. Item 18 ('How do you think your health status will be in the future?') was eliminated because it was not discriminant (ie, of the 5 response options, only the two extreme and positive ones, namely 'excellent' and 'good', were selected). As expected, the analysis of kurtosis and skewness evidenced high kurtosis (above 2) in items 7, 14, 16, 17, 37, 44, 45, 46, and 47, and high skewness (above 2) in items 16, 45, 46, and 47, in the direction of

respondents showing good health status after surgery. These items were not removed at this stage because such findings were expected and are consistent with the fact that the same item is used to assess pre- to post-treatment status.

3.2 | Analysis of factor structure of items comparing pre- to post-treatment status

The results of the exploratory factor analysis with the selected 24 items indicated a good fit of models with 5 (chi-square =210.71, P =.0108, RMSEA =0.067, 90% CI = [0.034, 0.093], CFI =0.977, TLI =0.961) and 6 (chi-square =170.85, P =.0868, RMSEA =0.052, 90% CI = [<0.001, 0.083], CFI =0.987, TLI =0.976) factors. The 6-factor solution was preferred because it allowed to differentiate factors as a function of pre-treatment (3 factors) and their corresponding post-treatment items (3 factors).

According to the exploratory analyses (Appendix 1), item distribution in the 5-factor solution should be factor 1 (items

1, 4, 9, 10, 13, 14, and 15), factor 2 (items 5 and 8 to 14), factor 3 (items 3, 4, and 7), factor 4 (items 2, 6, and 14), factor 5 (items 5 and 17 to 24), and factor 6 (items 15 and 16). This item distribution was problematic because factors included pre-treatment and post-treatment items together and because cross-loadings were frequent (eg, items 4, 5, and 15). Therefore, based on the results but grouping pre-treatment and post-treatment items in different factors, we proposed the following factor structure, which is described in Table 1: factor 1 (items 1, 4, 9, 10, 13, and 15, corresponding to 'mouth and language functioning and pain at pre-treatment'), factor 2 (items 5, 8, 11, 12, 14, and 16, corresponding to 'mouth and language functioning at post-treatment'), factor 3 (items 2 and 3, corresponding to 'breathing and hearing functioning at pre-treatment'), factor 4 (items 6 and 7, corresponding to 'breathing and hearing functioning at post-treatment', factor 5 (items 20, 21, 24, and 35, corresponding to 'psychosocial functioning at pre-treatment'), and factor 6 (items 22, 23, 25, and 36, corresponding to 'psychosocial functioning at post-treatment').

The confirmatory 6-factor model presented an excellent model fit according to CFI and TLI estimates and a good fit based on the RMSEA (chi-square = 325.46, P =.0001, RMSEA = 0.079, 90% CI = [0.056, 0.099], CFI = 0.954, TLI = 0.946). Additionally, as reported in Table 1, all items had high factor loadings in their corresponding scales (all ≥ 0.50). The modification indices only suggested that the model fit would slightly improve if item 5 ('At present, do you find difficulties in biting, eating and/or swallowing food?') was changed to the factors containing psychosocial functioning (factors 3a or 3b). This change, however, was not made to maintain the consistency and comparability of factors and because the model fit was already adequate and the improvement in fit after changing the item was very modest.

3.3 | Analysis of factor structure of items that do not have a pre-treatment and a post-treatment version

The results of the exploratory factor analysis indicated a good-to-excellent fit of models with 3 (chi-square = 280.59, P =.0100, RMSEA = 0.062, 90% CI = [0.032, 0.085], CFI = 0.958, TLI = 0.944), 4 (chi-square =232.98, P =.0955, RMSEA =0.047, 90% CI = [<0.001, 0.075], CFI = 0.978, TLI = 0.968), and 5 (chi-square = 193.92, P =.3117, RMSEA =0.028, 90% CI = [<0.001, 0.065], CFI = 0.993, TLI = 0.988) factors. As seen in Appendix 2, however, the model with 3 factors had an important number of cross-loadings with similar size of loadings. The 4-factor model had more acceptable number of cross-loadings. Item distribution, however, resulted in a factor

that was difficult to interpret (factor 2 combining items 27, 28, and 29 on physical appearance and items 40, 41, and 42 on family functioning).

A close look at the 5-factor model evidenced that these two groups of factors were better conceptualized in separate factors, but indicated that item 42 ('Does cleft management influence your /your family life financially?') no longer loaded in the same factor as arguably related items 40 ('Has your pathology management ever generated any family conflicts?') and 41 ('Has your pathology management changed any family habit?'). These results and the analysis of item content made us think that these items might have been too difficult to be interpreted and answered, especially by children and young adolescents. Thus, these three items were removed from further analyses. Before conducting the final confirmatory analysis, we eliminated 4 additional items: items 19 ('How would you define the general health status of your mouth and/or nose?') and 38 ('Are you optimistic about your future life?') that loaded on all factors; item 17 ('How would you define your present health status?') that loaded on a factor that measured a conceptually unrelated content (factor 4, corresponding to 'institutional barriers'); and item 37 ('Are you satisfied with your present life?') that loaded on factors 1 and 3, corresponding to 'psychosocial functioning' and 'family and peer support'.

After removing the former items, an exploratory analysis was made with the remaining 18 items and the 4-factor solution was again supported (chi-square = 83.33, P =.2143, RMSEA = 0.046, 90% CI = [<0.001, 0.090], CFI = 0.991, TLI = 0.984). The factor loading of item 46 ('Are they willing to help you in making these decisions?'), however, was greater than one and presented a negative residual variance, arguably because of its high correlation with item 45 ('Do friends/colleagues/parents support your choices in managing your pathology?') due to the wording of item 46. Item 46 was also removed.

Thus, the final confirmatory analysis with a 4-factor solution with 17 items, which is presented in Table 2, was as follows: factor 1 (items 26, 30, 31, 32, 33, 34, 43, and 44, corresponding to 'psychosocial functioning'), factor 2 (items 27, 28, and 29, corresponding to 'physical appearance'), factor 3 (items 39, 45, and 47, corresponding to 'family and peer support'), and factor 4 (items 48, 49, and 50, corresponding to 'institutional barriers'). This solution had good face validity and fit indices (chi-square = 152.47, P = .0079, RMSEA = 0.076, 90% CI = [0.041, 0.106], CFI = 0.964, TLI = 0.957).

3.4 | Utility of pre-post scales

To explore the utility of pre-post scales, we compared the scores in the 6 scales that belonged to the pre-treatment

and post-treatment comparison part of the questionnaire (Table 3). Specifically, we compared pre- to post-treatment 'mouth and language functioning and pain', pre- to post-treatment 'breathing and hearing functioning', and pre- to post-treatment 'psychosocial functioning'. A non-parametric test (ie, Wilcoxon) was conducted because an analysis of normality distribution of scores (Shapiro-Wilk test) indicated that normality assumption was violated in the majority of scales. The analyses indicated an improvement in 'mouth and language functioning and pain' ($z = -6.20$, $P < .001$, $d = -2.67$, $r = -0.80$), 'breathing and hearing functioning' ($z = -4.56$, $P < .001$, $d = -1.46$, $r = -0.59$), and 'psychosocial functioning' ($z = -4.85$, $P < .001$, $d = -1.60$, $r = -0.62$).

3.5 | Sources of criterion validity

Spearman's correlations as opposed to Pearson's correlations were performed due to the violation of normality assumptions. The analyses of sources of criterion validity (Table 4) revealed that the psychosocial scales (pre-treatment, post-treatment, and current functioning) and family and peer support scales were the most consistently associated with self-esteem (general, social, home, school, and total) and self-efficacy (social and total) measures. The strength of the correlations was generally between small and medium. The remaining QoL scales, namely 'mouth and language', 'breathing and hearing', 'appearance', and 'institutional barriers', were weakly and generally non-significantly associated with self-esteem and self-efficacy dimensions.

4 | DISCUSSION

This study aimed to adapt the QoLAdoCleft into Spanish, as well as to make some adaptations to the scale that would allow comparisons between pre- and post-scores. After splitting the questionnaire into two sets of items (24 corresponding to pre- to post-treatment changes and 26 referring to current status) and removing 9 problematic items, we obtained a psychometrically sound factor solution for the scale. The first set of items allows the comparison of pre- and post-treatment status into the following factors: 'mouth and language functioning and pain', 'breathing and hearing functioning', and

'psychosocial functioning'. With the second set of items, a 4-factor solution ('psychosocial functioning', 'physical appearance', 'family and peers support', and 'institutional barriers') was obtained after eliminating 9 problematic items. The analyses of pre- to post-treatment changes in scores and sources of criterion validity supported the factor structure and utility of the proposed adaptation of the questionnaire.

Living with CL/P can have an important impact on the QoL individuals, so the assessment of QOL constitutes a key goal in this population.[2] Several measures of general and specific cleft QoL measures exist. The QoLAdoCleft,[17] however, has the advantage of including items that assess not only the current status of the individual, but also a retrospective evaluation of the QoL before treatment. This is important because reconstruction surgeries that aim to improve the function of the mouth and face are frequent and the evaluation of pre- to post-treatment status provides a valuable insight into the perceived effectiveness of treatments.[2,8]

Given the importance of QoL in these patients, similar studies have been carried out to validate QoL measures to enable the assessment of this variable in the cleft population. For example, a validation of the COHIP-SF questionnaire was carried out by Agnew et al[8] for cleft patients with the same age range as the present study. The COHIP-SF is used to assess both positive and negative aspects of OHRQoL in children with varying oral condition by measuring oral health, functional well-being, and socio-emotional well-being. The study concluded that this instrument is valid and reliable for measuring OHRQoL in children with orofacial clefts. Similarly, Ranganathan et al[10] tested the reliability and validity of the Patient-Reported Outcomes Measurement Information System (PROMIS), which measures HRQoL, across clinical conditions, for children with CL/P, they tested construct validity with the Pediatric QoL. This study, however, was carried out in a younger population compared with the current study (5 years old). Both studies used general, not cleft-specific, QoL measures. Patjanasoontorn et al,[30] however, developed and tested the reliability of the THAICLEFT QoL questionnaire for families of children with CL/P on the basis of the KINDL questionnaire for measuring QoL in children with illness, and WHOQOL-BREV for the measurement of generic QoL and the Impact On Family Scale, but the resulting instrument measured psycho-economic

TABLE 3 Utility of the pre-post scales, as evaluated with the Wilcoxon signed rank test

Quality of life	Pre Mean (SD)	Post Mean (SD)	z	P	d	r
Mouth and language	1.61 (0.97)	0.74 (0.62)	−6.20	<0.001	−2.67	−0.80
Breathing and hearing	1.57 (1.09)	0.95 (0.89)	−4.56	<0.001	−1.46	−0.59
Psychosocial	1.33 (1.15)	0.83 (0.88)	−4.85	<0.001	−1.60	−0.62

TABLE 4 Sources of criterion validity of the quality of life in cleft scale

	Self-esteem					Self-efficacy			
Quality of life	General	Social	Home	School	Total	School	Social	Self	Total
Mouth and language pre	0.25	0.13	0.10	0.11	0.24	−0.15	−0.13	−0.30*	−0.26*
Mouth and language post	0.11	0.06	−0.03	0.15	0.12	−0.04	0.11	−0.06	0.01
Breathing and hearing pre	0.24	0.23	−0.10	−0.02	0.16	0.05	−0.13	−0.01	−0.06
Breathing and hearing post	0.08	0.04	−0.03	0.11	0.11	0.12	0.02	−0.02	0.07
Psychosocial pre	0.52***	0.54***	0.33*	0.42***	0.59**	−0.21	−0.56***	0.06	−0.34**
Psychosocial post	0.60***	0.48***	0.24	0.43***	0.54***	−0.20	−0.38**	0.14	−0.23
Psychosocial function	0.59***	0.61***	0.34**	0.47***	0.65***	−0.23	−0.47***	0.06	−0.33*
Appearance	0.37**	0.24	0.12	0.25	0.32*	−0.22	−0.29*	−0.06	−0.27*
Family and peer support	0.40**	0.42***	0.40**	0.55***	0.58***	−0.24	−0.44***	−0.12	−0.33**
Institutional barriers	0.27*	0.14	0.12	0.09	0.20	−0.13	−0.22	0.02	−0.17

***$P < .001$; **$P < .01$; *$P < .05$.

need of the families, not of the children. Given the limited number of validation studies into QoL scales in children and adolescents with cleft, the present study represents a step forward in the field.

As noted in Introduction, even though the QoLAdoCleft has the advantage of including pre-treatment and post-treatment items, the 3-factor structure (physical, psychological, and social health) proposed by the authors[17] combined pre-treatment, post-treatment, and general QoL status in the same factors. This was problematic as it would not allow for the calculation of pre- to post-treatment changes in QoL status. To solve this problem and to improve the utility of the scale, we selected the scale items that evaluated status before and after treatment. The structure was somewhat similar to the original, in the sense that physical items and psychological and social items were separated. The analyses, however, suggested that physical items should be divided into 'mouth and language functioning and pain' and 'breathing and hearing functioning', whereas psychological and social items should be grouped into a single 'psychosocial functioning' factor. Most importantly, an adequate model fit was obtained by grouping items into pre-treatment and post-treatment items. Our analyses of pre- to post-treatment differences suggest that our classification is useful to detect changes in QoL after treatment.

In addition to providing a more useful classification of pre- to post-treatment items by grouping them into separate factors, a factor analysis was performed for the remaining 26 items about general QoL status. One important finding was that our results suggested removing 9 of these items, either because they were not discriminant, too general and

associated with many other items, or not consistently associated with the expected factor. The latter made us think that some items might have been too difficult to interpret and respond to by children and young adolescents.

In relation to the proposed elimination of items, it is important to note that the scale was originally tested with older adolescents and young adults. Additionally, the authors proposed, but did not assess for model fit, the existence of 14 subfactors in the scale (4 for physical health, 5 for psychological health, and 5 for social health), which suggests a large variability in item content.[17] These two features might help understand why up to 9 items were eliminated in the present study (ie, due to the younger age of participants in the present study and because content in the questionnaire is in fact very diverse). Testing the utility of the scale in younger individuals, as done in the present study, is of great interest because of their increased psychosocial vulnerability.[18] Additionally, the existence of a reduced number of factors (10 in the present study as opposed to 14 in the original study) is also preferable for parsimony. Note that 9 items were eliminated, but this resulted in a reduction of 4 factors, which makes the questionnaire more parsimonious.

A final contribution of the study is the analysis of sources of construct validity, which was absent in the questionnaire development study.[17] Our results support the idea that the 'psychosocial' and the 'family and peers' scales are the ones presenting a more robust and strong association with several dimensions of self-esteem and self-efficacy. This finding is not new in patients with CL/P. Recently, a study conducted in Finland pointed out the relevant role of family and peer

support, along with good outcome, and oral health care being a normal routine, in motivation to attend a long and demanding oral health care regimen.[24] Interestingly, the more physical factors of QoL (eg, 'mouth and language', 'breathing and hearing', and 'appearance') and the 'institutional barriers' scale were generally unrelated or weakly associated with self-esteem and self-efficacy. These results provide further evidence for the need to evaluate QoL from a broad, multidimensional perspective, as well as to evaluate treatment success not only in terms of physical reparation, but also in relation to subjective psychosocial well-being.[1]

The present study also has some limitations. The first refers to the small number of participants. Even though the current sample size (n = 60) is larger than the one used for the questionnaire development (n = 40),[17] a formal sample size estimation was not conducted. A priori sample size calculations in validation studies are still infrequent (ie, <10% of studies), arguably because psychometrically sound and robust recommendations are still missing.[31] Although absolute rules for sample size calculations in validation studies do not exist, larger samples are preferable,[32] so a sample increase would be desirable in replication of the present study findings. It should be noted, however, that limited sample sizes are frequent in studies into CL/P.[23] In addition, a convenience sample, obtained from two specialized centers, was used, which limits the generalizability of the results. Another aspect that might negatively impact the generalizability of findings is that a percentage of potential participants were not willing to participate. Thus, it is possible that patients with poor or excellent quality of life were not interested in participating in the study and were not represented in our sample. Another study shortcoming refers to the fact that a qualitative interview was not conducted to evaluate why some items appeared to have a poor functioning. Thus, although we hypothesized that certain items (eg, impact of cleft on the family economy or the family habits) might have been difficult to answer by children and young adults, this is only speculative. Another aspect to consider is that the data on the temporal stability of items were not obtained, so this would be an interesting goal for further research. Finally, the retrospective assessment of patient status (especially in the pre-treatment moment) could lead to an overestimation bias of the current QoL compared with the previous one. Thus, evaluating patients in real time both before and after an intervention would be preferable when possible.

To conclude, the present study has important clinical implications. First, it provides a new factor structure for the QoLAdoCleft so that perceived CL/P changes after treatment can be assessed in three different QoL areas (ie, 'mouth and language functioning and pain', 'breathing and hearing functioning', and 'psychosocial functioning'). Second, it shows that the scale can be effectively used in children and adolescents and the number of subscales can be reduced from 14

to 10 by removing 9 items only. Finally, it shows that some scales are associated with important outcomes in children and adolescents with CL/P (ie, self-esteem and self-efficacy), which suggests that its application is likely to provide relevant and valid information.

WHY THIS PAPER IS IMPORTANT TO PEDIATRIC DENTISTRY?

- The validation of this questionnaire offers a coherent factorial structure that allows an assessment of the pre-post intervention perception of improvement, as well as presenting adequate indicators of validity and reliability.
- Paediatric dentists will be able to evaluate QoL in cleft patients at early ages, and to carry out personalized prevention measures.

ETHICS STATEMENT
This study was approved by the Ethics Committee of Rey Juan Carlos University (protocol code 110 720 166 716).

CONFLICT OF INTEREST
The authors declare no conflict of interest.

AUTHOR CONTRIBUTIONS
Ana Ruiz collected data and wrote the manuscript; **Carlos Suso** analyzed the data and drafted the initial manuscript; **Martín Romero** and **Carmen Gallardo** reviewed and revised the manuscript; and **Cecilia Peñacoba** conceptualized and designed the study. All authors reviewed the manuscript and approved the final manuscript.

DATA AVAILABILITY STATEMENT
The data underlying this article will be shared on reasonable request to the corresponding author.

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APPENDIX 1

Factor loadings of 5- to 6-factor models from the exploratory factor analysis

Original item number	5-factor solution					6-factor solution					
	1	2	3	4	5	1	2	3	4	5	6
1		0.39	0.40							0.50	
2	0.89						0.79				
3		0.95				0.87					
4		0.50		0.34		0.43				0.41	
5			0.45					0.33	0.37		
6	0.62						0.83				
7		0.89				0.96					
8			0.48						0.46		
9			0.71						0.53	0.35	
10			0.80						0.39	0.66	
11	−0.34		0.68						0.85		
12			0.90						0.86		
13			0.78							0.74	
14	0.34		0.87				0.44		0.55	0.42	
15			0.34							0.74	0.37
16					0.91						0.93
20				0.91				0.90			
21				0.96				0.95			
22				0.82				0.80			
23				0.88	0.38			0.88			0.39
24				0.74	−0.33			0.75	0.32		
25				0.76				0.75			
35				0.86				0.85			
36				0.69				0.68			

APPENDIX 2

Factor loadings of 3- to 5-factor models from the exploratory factor analysis with items that do not have a pre-treatment and a post-treatment version

Original item number	3-factor solution			4-factor solution				5-factor solution				
	1	2	3	1	2	3	4	1	2	3	4	5
17			0.34				0.35				0.32	
19								0.33				
26	0.56		0.40	0.53			0.33	0.39	0.43			
27	0.59		0.49		0.73			0.92				
28	0.60				0.56		0.41	0.77				
29	0.54				0.55			0.54				
30	0.80			0.52	0.46			0.53	0.37			
31	0.93			0.76				0.47	0.62			
32	0.77			0.69				0.43	0.58			
33	0.60			0.51					0.48			0.34
34	0.64			0.76					0.73			
37	0.47	0.55		0.42		0.51		0.35	0.32	0.51		
38		0.33								0.36		
39		0.49				0.49			0.32	0.47		
40	0.37	0.54			0.82							0.78
41	0.43	0.42			0.75			0.32				0.85
42					0.42							
43	0.82			0.89					0.88			
44	0.69			0.63					0.57			
45		0.95				0.94				0.94		
46		0.93				0.94				0.95		
47	0.38	0.63		0.42		0.62			0.33	0.65		
48			0.88				0.90				0.87	
49			0.91				0.91				0.96	
50			0.98				0.98				0.92	